Potential Adjunctive Role of Radiosynovectomy in Primary Synovial Osteochondromatosis of the Knee: A Case Report

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Abstract Primary synovial osteochondromatosis (PSOC) is a rare but clinically significant cause of morbidity especially in the male population. Surgery is the primary treatment of choice, but the recurrence rate is reported to be high. Moreover, the presence of widespread loose bodies makes it a cumbersome procedure. The complete removal of the disease is tough at times and results in early recurrence. Radiosynovectomy is an established technique for treating various joint arthropathies. The role of radiosynovectomy in case of PSOC has not yet been explored. This case report described the case of a young male with PSOC of the knee joint who was treated with radiosynovectomy for pain relief. The patient reported complete relief from the pain along with significant improvement in joint mobility. The post-therapy three-phase bone scan also validated the reduction in joint inflammation. The patient was taken for surgical removal of the redundant loose bodies after a significant improvement in the pain and reduction in inflammation. Post-therapy radiation fibrosis of the synovium also helped in the en bloc removal of the disease. The role of radiosynovectomy in PSOC needs to be further explored concerning its potential role as an adjuvant to surgical procedures.

Keywords Primary synovial osteochondromatosis (PSOC) · Radiation synovectomy · Radiosynoviorthesis · Rhenium-188 (¹⁸⁸Re)

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Introduction

Synovial chondromatosis is a rare benign pathology characterized by the formation of multiple cartilaginous nodules or osseous bodies in the involved joints and less commonly in the tendon sheaths or bursae. Though synovial chondromatosis can be secondary to underlying arthritis or degenerative joint disease, it often occurs in an otherwise normal joint, described as PSOC. First described in 1833 by Leanacc, it is a rare disease entity with an incidence of only 1:100,000 [1]. The disease has a male predominance and usually affects young men in the 3rd–5th decades. It is often monoarticular involving the knee in the majority of cases (~50%) followed by the hip, elbow and the shoulder joints in decreasing order of frequency [2]. Radiosynovectomy or radiosynoviorthesis is an established technique involving intra-articular administration of radiopharmaceuticals labeled with high-energy beta-emitting radionuclides for the treatment of various joint pathologies including rheumatoid arthritis and other inflammatory arthropathies [3]. We present the first case of a young male with PSOC of the right knee with severe joint immobility and pain treated with radiosynovectomy prior to surgery, facilitating en bloc disease resection with adjunctive therapeutic benefit.

Case Report

A 25-year-old male patient presented with progressive swelling and pain around the right knee joint for the last 1.5 years. The history was negative for any previous trauma or other articular symptoms. There was large peri-articular swelling of the right knee with associated tenderness and restriction of joint mobility on examination. Evaluation with plain radiography revealed multiple irregular calcific deposits in the peri-articular soft tissues of the right knee (Fig. 1a). Magnetic resonance imaging

Fig. 1 **a** Baseline plain radiograph of the right knee joint showing multiple calcified nodular lesions in relation to the supracondylar region of the right femur. **b** Photograph showing the en bloc resected surgical specimen with a hard nodular surface. **c** Radiograph of the knee joint following surgery depicting adequate removal of the diseased synovium (with surgical clips in situ)

demonstrated diffuse synovial thickening (involving the peri-articular bursal recesses) and associated moderate synovial effusion. Numerous hypointense focal areas were also noted in the synovium and bursal recesses suggesting a diagnosis of synovial chondromatosis. Synovial fluid evaluation was non-contributory. The patient was referred for the management of pain and swelling with a radiosynovectomy procedure. Three-phase ^{99m}Tc -MDP skeletal scintigraphy of the knee region was performed

as a prerequisite for radiosynovectomy. It revealed increased flow and peri-articular soft tissue pooling of the tracer suggesting active synovial inflammation in the right knee (Fig. 2a–c). Radiosynovectomy was performed by intra-articular injection of 555 MBq of ^{188}Re -tin colloid prepared in the in-house radiopharmacy. Synovial fluid was initially aspirated followed by instillation of the radiopharmaceutical into the joint with a saline flush to clear any residual activity in the same sitting

Fig. 2 **a–c** Three-phase skeletal scintigraphy of the knee region depicting increased tracer distribution (arrow and arrowheads) in the right knee region in all the three phases (perfusion, blood pool and delayed). **d–f**

Significant reduction (arrow and arrowheads) in the tracer distribution (all the phases) is noted in the follow-up three-phase scintigraphy at 6 months

Table 1 Clinical and imaging parameters

Clinical/imaging parameters	Pre-therapy	Post-therapy (3 months)	Post therapy (6 months)
1. Pain	Severe (even at rest)	Decreased up to 70%	Complete resolution
2. Tenderness	Severe	Mild	No
3. Knee circumference at the level of the patella	40 cm	36 cm	34 cm
4. Angle of flexion at the knee joint	0–10°	90°	120–130°
5. Three-phase skeletal scintigraphy at pre-therapy and at 3 months post-therapy			
Flow	Increased	-	No
Soft tissue pooling	Present (significant)	-	Minimal
Delayed	Increased	-	Increased

under strict aseptic conditions. The joint was briefly mobilized to ensure uniform distribution of the radiopharmaceutical within the synovium and then immobilized for 48 h using a knee splint to minimize extra-articular spread. Post-therapy scintigraphy at 1 h revealed adequate homogeneous intra-articular distribution of the radiopharmaceutical. Subsequently, the patient was followed up at 3- and 6-month intervals post-therapy with clinical examination demonstrating remarkable improvement in the form of complete resolution of pain, significant reduction in swelling and restoration of joint mobility. A three-phase skeletal scintigraphy performed at 6 months showed a significant reduction in the bulk and inflammatory activity (Fig. 2d–f, Table 1). The patient underwent surgical synovectomy in view of the residual swelling and limitation of mobility. The resected surgical specimen was hard with a nodular surface and showed variable-sized whitish nodules on cut sections. Histopathology revealed areas of loose myxomatous connective tissue with multiple lobular clusters of chondrocytes (forming hyaline loose bodies) on a background of dense hyalinization and calcification confirming the diagnosis of synovial chondromatosis. There was a noticeable finding of the substantial radiation-induced dense hyalinization in the pathological synovium, which facilitated the en bloc resection (Fig. 1b). The subsequent radiograph revealed adequate removal of the involved synovium (Fig. 1c). The patient is currently asymptomatic with complete resolution of pain and near complete restoration of joint mobility.

Discussion

The clinical presentation of PSOC often involves pain, swelling, stiffness and locking of the knee with reduced joint mobility, tenderness, palpable loose bodies and crepitus noted on physical examination. Secondary osteoarthritic changes in long-standing cases can result in severe disability [4]. The debate is still ongoing regarding the pathophysiology (synovial metaplasia vs. benign neoplasm), but recent cytogenetic studies have favored a benign neoplastic etiology [5], although the rare transformation into synovial chondrosarcoma is also reported in the literature [6]. Milligram proposed three

phases of disease progression: the early phase having an active intra-synovial disease without any loose bodies, the transitional phase with both active synovial disease and loose bodies, and the late stage with multiple loose bodies without any intra-synovial disease [7]. Surgery, either open or arthroscopic excision, is the treatment of choice. However, a high recurrence rate (of even up to ~23%) is reported in the literature [8].

Different radiopharmaceuticals in colloidal form are used for radiosynovectomy depending upon the joint size [3]. The radionuclide used in this index case was (^{188}Re). It has certain advantages such as having both beta and gamma radiation, around the clock availability of a $^{188}\text{W}/^{188}\text{Re}$ generator and ease of in-house Re-188 tin colloid preparation, making it a very cost-effective treatment option. It is often employed as an alternative therapy to early surgical synovectomy or as an adjunctive treatment to surgical procedures for optimal results [9]. To our knowledge, the utility of radiosynovectomy in the management of PSOC has not been reported so far in the existing literature. Complete removal of this disease is warranted in view of the possibility of post-surgical recurrence and the rare but plausible risk of malignant transformation. In the current patient, radiosynovectomy yielded dramatic improvement of symptoms, alleviating the need for analgesics, and also facilitated effortless intraoperative en bloc resection of the diseased synovium (unlike the conventional cumbersome piecemeal technique). The present case shows the potential role of radiosynovectomy in the management of synovial chondromatosis as an adjunctive therapeutic intervention prior to surgery.

Compliance with ethical standards

Conflict of interest Shelvin Kumar Vadi, Devendra Kumar Chouhan, Arun Kumar Reddy Gorla, Jaya Shukla, Ashwani Sood and Bhagwant Rai Mittal declare that they have no conflict of interest. There is no source of funding.

Ethical approval All procedures performed in studies involving human participants were in accordance with Declaration of Helsinki and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from participants included in the study.

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